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RESEARCH ARTICLE

Baseline Features Influencing the Effectiveness of Retraining Therapy for Writer's Cramp

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Abstract: **Background:** The effectiveness of retraining therapy (RT) for writer's cramp is difficult to predict and its determinants are unknown. **Methods:** We examined factors potentially predicting improved legibility after RT in patients with writer's cramp (WC). We reviewed the files of 693 WC patients treated with RT from 1995 to 2009. Standardized assessments were made both at baseline and after 2 months of RT in 305 patients. The effect of RT on legibility was evaluated by using the handwriting subscore of the Burke-Fahn-Marsden (BFM) disability scale. Initial and final handwriting samples were blindly scored in random order. Associations between WC patterns and changes in legibility were identified by uni- and multivariable analyses. **Results:** Legibility improved by ≥1 point in the BFM handwriting subscore in 93 patients (31%). WC patients who improved were more likely to have synergic dystonic patterns involving the wrist and forearm (60% vs. 40%; $P = 0.03$) and less likely to have flexion of fingers F3 to F5 (19% vs. 81%; $P = 0.017$). Outcome was not related to gender, age, or dystonia duration. Our results confirm that retraining therapy could improve legibility in patients with writer's cramp. **Conclusions:** The pattern of writer's cramp can help to identify patients who are most likely to benefit from retraining therapy, regardless of age, gender, and disease duration.

Writer's cramp (WC) is a task-specific focal dystonia associated with abnormal movements and posture of the hand during writing. Legibility and speed are the main elements of handwriting performance. Legibility is the standard by which handwriting is judged by the writer and the reader, both professionally and socially.

Most studies of retraining therapy (RT) in this setting have focused on correction of the abnormal posture, with variable improvement.[1] Only a few small studies have examined the effect of RT on handwriting legibility in WC patients. Studies of various RT approaches did not take into account the possible influence of the pattern of dystonia (e.g., pronation and wrist or finger involvement) on the response to treatment.[2] Yet, it is likely that the response to RT will vary according to the WC clinical pattern, as observed in patients treated with botulinum toxin (BoNT).[3] Therefore, we examined baseline factors potentially influencing the response of WC patients to RT, in terms of handwriting legibility. We blindly rated the legibility of pre- and post-RT standardized handwriting samples in 305 WC patients who received standardized RT between 1995 and 2009.

Methods

Subjects

From a population of 693 consecutive adult WC patients examined in the Department of Physiotherapy, Sainte Anne Hospital (Paris, France) from January 1995 to December 2009, we

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reviewed the medical records of those who had at least 2 months of RT. All the patients were diagnosed and referred by specialists in movement disorders (i.e., task-specific dystonia during writing). WC (abnormal postures and alteration of handwriting) manifests itself during a handwriting sample of a sentence. BoNT injections were not used with the retraining therapy. The patients did not receive any other treatment during the RT period.

Retraining Therapy

Each fortnight, all the patients underwent:

1. An individual 45-minute session of motor sequence learning[4] and task-oriented training[5] toward handwriting[6] managed by the physiotherapist (PT), including:
   - **(i)** Exercises for loss of dexterity (finger agility, finger dissociation, and fine motor coordination)
   - **(ii)** Strengthening exercises to correct poor posture and adjust muscle imbalance as well as reinforcement of the small muscles of the hand used in holding a pen and writing
   - **(iii)** Prewriting exercises: an intensive program of precision grip training; graphic finger training with specific graphic exercises chosen to avoid reproducing the situation, which triggered WC (e.g., drawing exercises)
   - **(iv)** Writing exercises: Patients were trained to maintain an ergonomic pen-hold, postural control and kinesthetic finger adjustment, control of low-level pressure on the pen, to adjust pen angle and control the speed during increasingly complex handwriting exercises.

2. A daily half-hour home retraining regime prepared by the PT, including:
   - (i) Precise manipulative movements, as well as repetitive and effortless writing exercises.

The team of full-time permanent PTs remained the same throughout the study period. We analyzed the specific effect of PT-driven therapy. The consistency of the RT was assured by a short-term session. The RT was mainly aimed at modifying the abnormal dystonic movement and gradually restoring normal writing patterns.

Study Design

We collected data on demographics, handedness, and WC duration. Evaluations were performed before and after the end of RT. The quality of handwriting was evaluated in terms of its legibility.

At the first visit, patients had a standardized clinical evaluation, including provision of a standardized handwriting sample ("*je respire le doux parfum des fleurs*" ["*I smell the sweet scent of flowers*"]). We focused on the dystonia pattern, WC type (tonic with abnormal posture, mobile with abnormal movements ± superimposed myoclonus or tremor, and writing tremor) and the presence of abnormal joint mobility (hyperlaxity or stiffness). We considered separately forearm, wrist, and finger abnormal patterns and also analyzed complex synergic movements (synergic patterns) of the wrist and forearm (combination of pronation or supination, flexion, or extension) and of the hand (pinch: first and second digit; grasps: all digits or third to fifth digits). At the end of the RT regime, patients were asked to write the same sample sentence as previously. To assess the legibility for each sample of handwriting, one single score was obtained in consensus by two raters (J.F.B. and L.S.L.) who were blind to the patient's condition (i.e., before or after RT). The initial and final handwriting samples were anonymized for each patient and were presented in random order. The raters used the Burke-Fahn-Marsden (BFM) disability scale handwriting subscore (0: normal; 1: slight difficulty, legible; 2: almost illegible; 3: illegible; 4: unable to grasp to maintain hold of the pen).[7] A score of 4 was obtained from the initial records, from the standardized clinical assessment. Improvement was defined as a ≥1-point before-after decrease in the BFM subscore.

According to the French legislation in force at the time of the study, we did not require ethics approval or written informed consent, given that the study consisted of a retrospective analysis of anonymous data collected prospectively as part of routine clinical care.

Statistical Analyses

Continuous variables were expressed as the mean (standard deviation; SD) or median (interquartile range; IQR) and categorical variables as percentages. Continuous variables were compared using a *t* test or Mann-Whitney's U test, and categorical variables were compared using the Pearson's chi-squared test or Fisher's exact test, as appropriate. Crude and adjusted odds ratios (ORs) were calculated in logistic regression models.

Results

Overall, 378 of 693 patients examined met the inclusion criteria (Fig. 1). Ineligible patients did not differ significantly from eligible patients in terms of age, gender, disease duration, or type of dystonia (data not shown). We excluded, respectively, 26 and 47 patients for whom no standardized writing sample was available at the initial or final visit. Characteristics of the remaining 305 patients are summarized in (Table 1).

Median duration of RT was 196 days (IQR, 234) with a median number of sessions of five (IQR, 8).

After RT, 93 (31%) patients had an improvement in handwriting legibility. The dystonia pattern was the key factor influencing the outcome of RT: Patients who improved were more likely to have a synergic pattern involving the wrist and forearm (60% vs. 40%; $P = 0.03$) than those who did not improve. Patients who had flexion of the third, fourth, and fifth fingers (F3–F5) were less likely than other patients to improve (19% vs. 81%; $P = 0.017$). Both the relation between "improvement" and "baseline wrist and forearm synergic pattern" (OR = 1.7; 95% confidence interval [CI]: 1.0–2.9;

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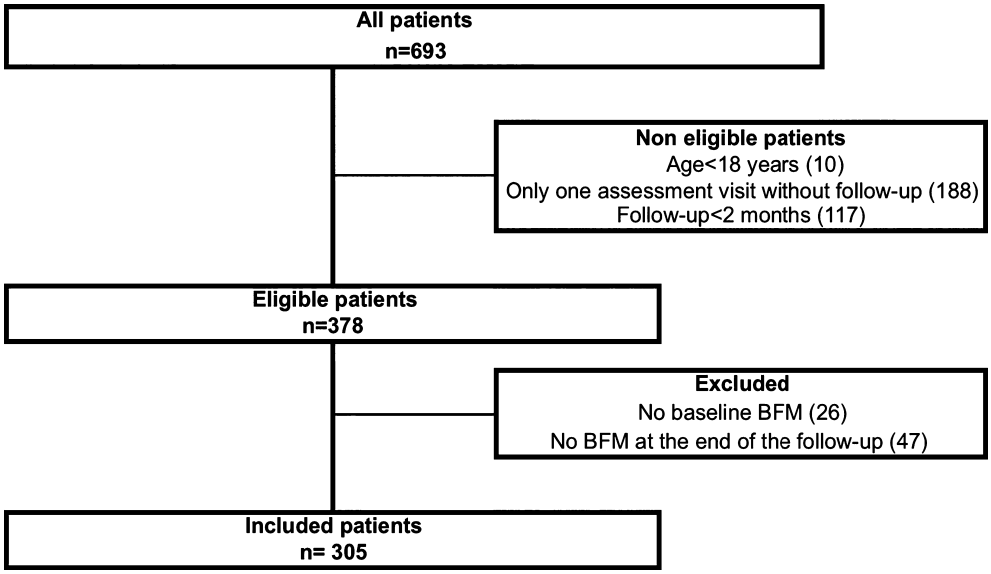

Figure 1 Flow chart of the study, eligibility of the patients.

$P = 0.05$) and the relation between "little or no improvement" and "baseline flexion of fingers F3 to F5" (OR = 0.5; 95% CI: 0.2–0.9; $P = 0.03$) persisted after adjustment for age and gender. Further adjustment for disease duration did not affect the results (synergic: OR = 2.2; 95% CI: 1.1–4.3; F3–F5: OR = 0.6; 95% CI: 0.3–1.3), and neither did the dystonia pattern of the forearm, wrist, or fingers (not shown).

Discussion

The main objective of the task-oriented RT program for patients with WC was to improve the writing pattern and fluidity of movement while holding the pen, in order to increase the legibility of the patient's handwriting. Our evaluation focused on the visible trace, rather than the writing gesture, given that it is the most important factor in professional and social interactions. We blindly rated the legibility of pre- and post-RT standardized handwriting samples presented in random order, using the handwriting subscore of the validated BFM disability scale.

We found that 31% of WC patients had an improvement in legibility after at least 2 months of standardized RT. The baseline pattern of dystonia was predictive of the outcome of RT: WC involving the wrist and forearm with a synergic pattern was more likely that other patterns to improve, independently of age, gender, and disease duration. Patients with such a pattern of WC should thus be encouraged to join an RT program, whatever their age at onset and their disease duration. Our findings are important for clinical practice given that they could help clinicians to identify patients who are most likely to benefit from RT, thereby improving the selection of the most appropriate therapeutic strategy.

The strengths of our study include the very large number of patients (n = 305), as compared to previous studies,[8] the single-center design ensuring consistency in the clinical description and evaluation of WC, and the standardization of the RT program (2 months of training sessions every fortnight by the PT and of daily self-training between these PT sessions), and the use of a standardized evaluation of motor performance (handwriting legibility on the BFM disability scale) by examiners who were unaware of the patient's pre- or post-RT status. Limitations of this study include its retrospective design, the lack of long-term evaluation, and the use of a BFM disability subscore that may not be sensitive enough to identify slight changes in legibility and might thus underestimate the impact of RT. Because we performed multiple analyses, one might argue that our findings may be owing to chance and that we should have adjusted our tests for multiple comparisons. However, we consider that our analyses were mainly exploratory, a situation for which it is generally accepted that multiple test adjustment is difficult and not required.

The proportion of our patients who improved after RT (31%) is very similar to that observed in a previous, smaller study (50 patients), in which a significant improvement in handwriting performance was found in one third of patients after brief RT (six sessions at variable intervals over 4 months).[9] In this study, several different aspects of handwriting, such as letter size and handwriting velocity, were evaluated with a digitizing tablet. Although different methods of evaluation were used, the efficacy of the handwriting training was equally visible (better handwriting performance) by using either a "simple technique" (blind evaluation of legibility as in our study) or a more "sophisticated" one (kinematic handwriting analysis).[9] Moreover, the predictive value of wrist and forearm synergy is consistent with the findings of a study of factors associated with improvement of motor disability after BoNT injection.[3] Indeed, the WC patients who improved most had a pronation/flexion pattern. It is therefore necessary to take account of the dynamic networks linking the forearm to the wrist, hand, and fingers when treating WC.

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TABLE 1 Characteristics of the study population and determinants of the response to retraining therapy

Characteristics	All Patients (n = 305)	Improvement (%)		*P* Value
		Yes (n = 93)	No (n = 212)	
Patients				
Age at first evaluation, mean (SD) years	43.2	43	43.6	0.70
Gender				
Male	124	39	85	0.76
Female	181	54	127	
Dominant hand				
Right	275	82	193	0.29
Left	25	10	15	
Joint abnormality	60	18	42	0.93
Dystonia				
Disease duration (yr)				
<1	13	3	10	0.17
1 to 3	48	12	36	
3 to 5	43	14	29	
5 to 10	30	15	15	
>10	70	20	50	
Dystonia type				
Tonic (abnormal posture)	210	65	145	0.43
Mobile (abnormal movements or posture+myoclonus/tremor)	57	14	43	
Writing tremor	38	14	24	
Shoulder involvement	25	7	18	0.78
Forearm involvement				
No	166	45	121	0.28
Pronation	111	40	71	
Supination	28	8	20	
Wrist involvement				
No	137	38	99	0.68
Extension	70	23	47	
Flexion	81	26	55	
Ulnar flexion	20	10	10	
Radial flexion	1	0	1	
Elementary finger movements				
Flexion or extension of F1/F2	155	43	112	0.29
Flexion of F1 to F5	41	10	31	0.36
Synergic finger patterns				
Flexion of F1/F2 (pinch)	123	34	89	0.37
Flexion of F3 to F5 (grasp)	69	13	56	0.017[a]
Synergic patterns[b]				
Any synergic movement of the wrist and forearm[b]	80	32	48	0.03[c]

[a]Associated with little or no improvement.
[b]Synergic movement of wrist and forearm: any of the following patterns (pronation+wrist flexion; pronation+wrist extension; ulnar inclination [ulnar adduction]+wrist flexion; ulnar inclination+wrist extension; radial inclination+wrist extension).
[c]Associated with improvement.

Wrist orientation can strongly influence grasp-related activities.[10] Coordination of intrinsic and extrinsic hand muscles is dependent on the wrist joint angle during two-digit grasping.[11] Commands involving proximal muscles (upper-limb position) influence the activity of distal muscles required for grasping, such as the hand configuration during handwriting.[10] Therefore, in patients with a pronation/flexion pattern, a better motor command for grasping, adapted to the writing gesture, would be obtained through RT corrective exercises. Indeed, the exercises target a more functional position or motion of the forearm and wrist joints. Likewise, it appears that the muscular overflow corresponding to an excessive response of the muscles naturally involved in the functional synergic writing pattern (forearm and wrist muscles) may benefit more from RT than those not involved in the normal writing gesture, such as flexion of F3 to F5. The mechanisms underlying the effects of RT, especially regarding the dystonia pattern, are poorly known. RT might act on loss of inhibition, sensorimotor integration, cortical plasticity, or cerebellar network activity.[12] If modulation of plasticity is the main substrate of the beneficial effect of RT, our study suggests that it is not influenced by age or by the duration of WC. These findings may constitute a preliminary background for studies exploring the dynamics of network modulation associated with retraining therapy.

Author Roles

(1) Research Project: A. Conception, B. Organization, C. Execution; D. Supervision; (2) Statistical Analysis: A. Design, B. Execution, C. Review and Critique; (3) Manuscript: A. Writing of the First Draft, B. Review and Critique.

J.-P.B.: 1A, 1B, 1D, 2A, 2B, 2C, 3A

E.T.: 1A, 1B, 1D, 2A, 2B, 2C, 3A

L.S.R.: 1C

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